Thorax (1968), **23**, 69.

Endobronchial teratoma associated with bronchiectasis and bronchiolectasis

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A teratoma in a young West Indian of Negro race is reported. The teratoma presented radiologically in the left upper lobe as an ill-defined shadow which contained a crescent-shaped translucent area and simulated a mycetoma. In addition, the left lung showed widespread nodular shadows. The left lung was resected and the teratoma was found to be endobronchial in position. This is a very rare site for a teratoma as only one of the 15 previously reported intrapulmonary teratomata may have been endobronchial. The remainder of the left lung remote from the tumour showed generalized bronchiectasis both radiologically and pathologically. The bronchiectasis was of follicular type and in addition there was widespread bronchiolectasis. The inflammatory reaction associated with the latter was responsible for the nodular shadows. The significance of these changes in relation to the teratoma is discussed.

The majority of intrathoracic teratomata are found in the mediastinum (Morrison, 1958; Le Roux, 1960) and those which occur in the lungs are among the rarest of tumours (Spencer, 1962). Ali and Wong (1964) reported a case of intrapulmonary teratoma, and in their review of the literature found 14 previously published cases. A further case of intrapulmonary teratoma was reported by Trivedi, Mehta, and Nanavaty (1966). Another unusual intrathoracic site is the pericardial cavity, and Adler, Taheri, and Weintraub (1960) and Kalter (1961) have reported a cystic teratoma in this position.

In view of the rarity of intrathoracic teratomata which lie outside the mediastinum, the authors wish to report a teratoma which was endobronchial in position. A search of the literature has failed to reveal any similar case with the possible exception of that described by Laffitte (1937). The present case was also interesting because of the confusing radiological presentation. In addition, all the bronchi of the lung which contained the teratoma showed bronchiectatic and bronchiolectatic changes of an unusual type.

CASE REPORT

A male Negro carpenter from St. Kitts, Nevis, gave a history of intermittent left pleuritic pain with a productive cough for five years and recurrent haemoptyses for three years prior to his admission to the University Hospital of the West Indies on 3 May 1966.

In April 1964 an exploratory thoracotomy had been performed in Puerto Rico for an undiagnosed opacity in the left upper lobe. An inflammatory mass was found and histological examination of several biopsy specimens revealed inflammation without any evidence of neoplasm. Post-operatively an empyema developed which gradually resolved after drainage. His symptoms as well as the opacity in the left upper lobe persisted.

On physical examination his general condition was good and the abnormal signs were confined to the left side of the chest. The anterior chest wall was slightly flattened, chest movement was restricted, and there were diminished breath sounds and a dull percussion note over the left lower lobe area. The haemoglobin was 12 g./100 ml., and the white cell count was 7,600/c.mm. with a normal differential count. Sputum examination for acid-fast bacilli, fungi, and malignant cells was repeatedly negative.

Radiographic examination of the chest (Fig. 1) revealed a mass with a somewhat lobulated and irregular margin. The shadow of the mass merged with that of the left hilum. Two small metal clips from the previous thoracotomy were shown. Tomography demonstrated the irregularity of the peripheral margin of the mass more clearly and also the presence of a crescent-shaped translucent area in its upper pole (Fig. 2). Because of the translucent area the mass was thought to be a mycetoma. In addition, nodular shadows were also demonstrated throughout the left lung. They were large and measured up to 5 cm. in diameter (Fig. 3).

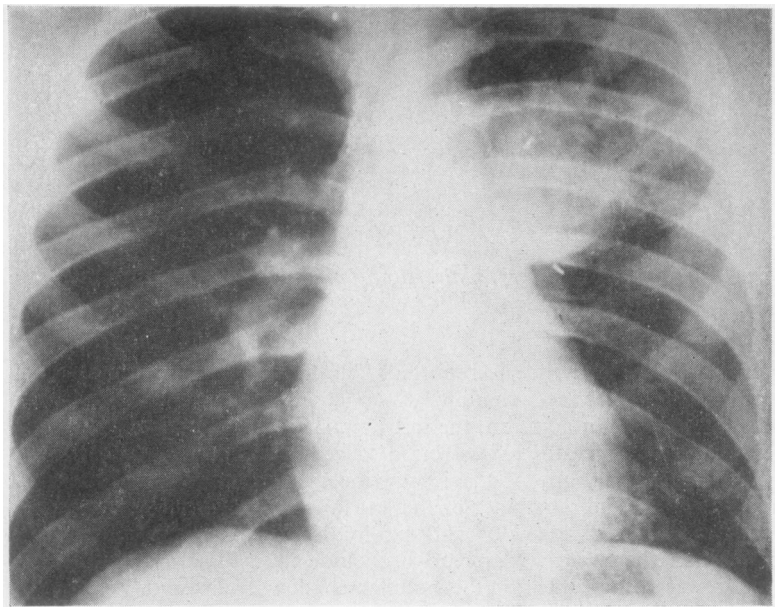


FIG. 1. *Postero-anterior radiograph of the chest showing the mass with a lobulated and irregular margin in the left upper lobe.*

Bronchography was performed by percutaneous puncture of the cricothyroid membrane and the injection of 15 ml. of propyliodone (Dionosil Oily) into each lung (Fig. 4). The right bronchial tree was normal. On the left side, bronchi in the neighbourhood of the mass were distorted, with narrowed and dilated segments. These changes were compatible with pressure from the adjacent tumour. The bronchi of the lateral segment of the lateral lingular lobe were fairly normal in calibre but showed a beaded appearance in places. There was simple cylindrical bronchiectasis of the medial segment of the lingula. The apical segment of the lower lobe showed similar change to the lateral segment of the lingula, and the

FIG. 2. *Tomogram of the left upper lobe demonstrating the presence of a crescent-shaped translucent area within the mass.*

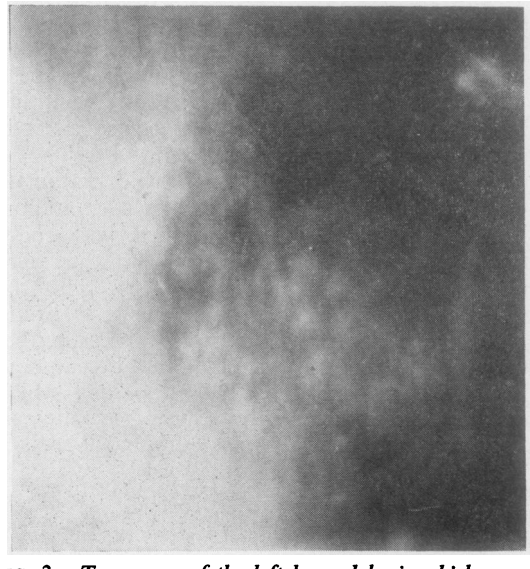


FIG. 3. *Tomogram of the left lower lobe in which many nodular shadows can be seen.*

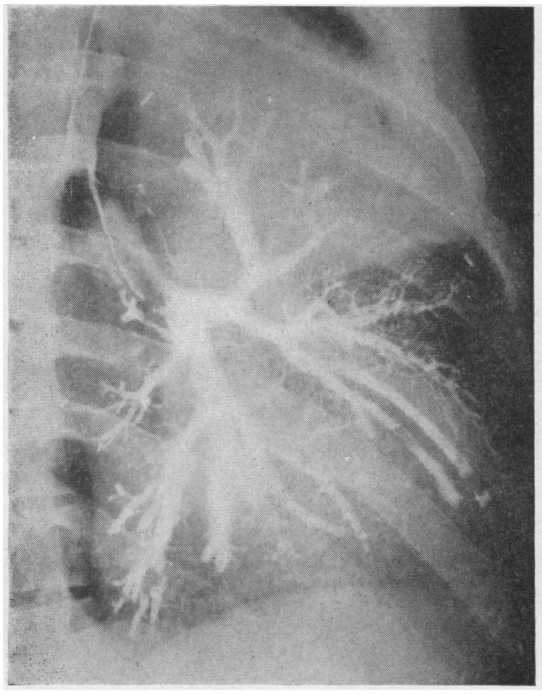


FIG. 4. *Bronchogram of the left lung with irregularity of the upper lobe bronchi in the region of the mass and bronchiectasis of the remaining bronchi.*

basal segments of the lower lobe showed a severe degree of cylindrical bronchiectasis. Therefore, all the bronchi of the left lung not directly affected by the tumour showed evidence of some type of bronchial abnormality.

Bronchoscopic examination showed mucoid secretions in the left upper lobe bronchus which was narrowed and displaced posteriorly by an extraluminal mass. A bronchial biopsy from the narrowed segment showed a considerably thickened epithelium and cellular atypia.

On 16 June 1966 a left postero-lateral thoracotomy was performed. The pleural cavity was completely obliterated by dense adhesions. There was a mass in the anterior segment of the left upper lobe, which was very adherent to all adjacent structures. A left pneumonectomy was performed because of the persistent haemoptyses and concomitant bronchiectasis of the lower lobe. Post-operative recovery was uneventful and he returned to St. Kitts on 15 July 1966.

The removed lung showed diffuse inflammation of the surface, which was covered by ragged skeins of fibrin. On the anterior margin of the upper lobe immediately below the apex there was a firm, rounded mass, 6 cm. in diameter, covered by dense pleural fibrosis (Fig. 5a). The parenchyma was firm throughout both lower lobes and cut with difficulty. All the bronchi in the upper lobe were dilated, including those which were remote from the mass. The latter was a polypoid lesion completely enclosed by the distended subapical bronchus, which formed a cavity, to the anterior wall of which the mass was attached.

The divisions of the bronchus distal to the mass were all markedly dilated. The mass, which measured $6 \times 4 \times 3$ cm., was composed of fleshy yellow and white tissue resembling brain in some areas. It had a thick, white, warty surface resembling that of the tongue, although numerous hairs projected from its posterior surface (Fig. 5b). The thin layer of lung between the mass and the pleura was compressed and largely replaced by cystic cavities filled by gelatinous material. The remaining bronchi were dilated, as demonstrated on bronchography, and the interlobular septa were prominent. Within almost every secondary lobule (Miller, 1947) there were several small yellow-white stellate areas which appeared to be centred about dilated terminal bronchioles (Fig. 5c).

On microscopical examination the polypoid mass shows the typical features of a benign teratoma with skin and essential appendages, primitive bronchial cartilage covered by respiratory type epithelium (Fig. 6a), intestinal epithelium with Brunner's glands, and pancreatic tissue containing islets of Langerhans (Fig. 6b). Each of the stellate areas shows an inflammatory reaction which is centred on a terminal bronchiole and extends along the respiratory bronchioles to the atrial ducts. The walls of the bronchioles show dense infiltrates of lymphocytes and plasma cells, with the formation of numerous germinal centres. Muscle bundles are prominent but there is little increase in fibrous or elastic tissue. In many areas adjacent alveolar walls are thickened by the dense inflammatory exudate and are lined by cubical epithelium (Fig. 6c). The arteries show no change, although a few of the arterioles lying within the lesions show intimal fibrosis. Muscular hypertrophy in the media is rarely seen.

DISCUSSION

The features of this case will be discussed under two headings—the endobronchial teratoma and the bronchiectatic changes.

THE ENDOBRONCHIAL TERATOMA The clinical presentation of haemoptysis for three years is not uncommon in patients with endobronchial tumours or bronchiectasis, and could have been due to either condition in this case.

The radiological appearances are interesting and should be discussed because of the incorrect diagnosis before thoracotomy. The presence of the crescent-shaped translucent area within the mass

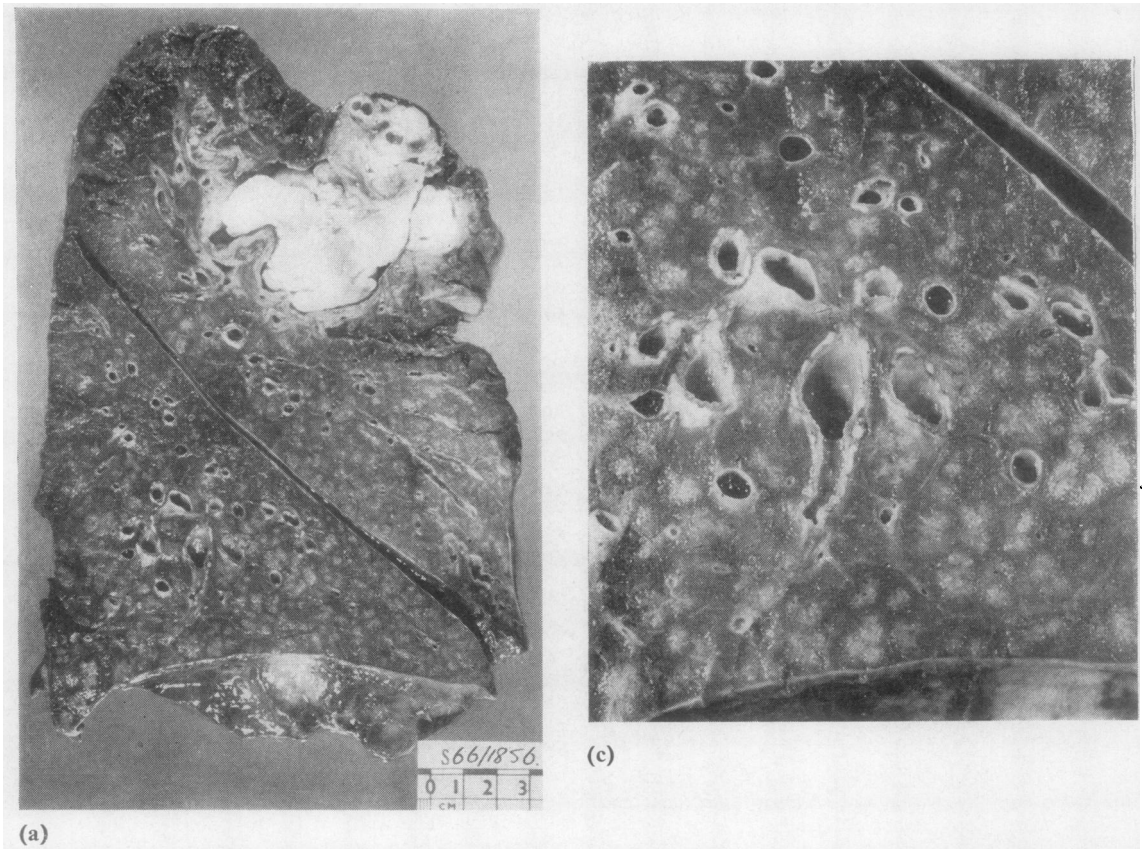
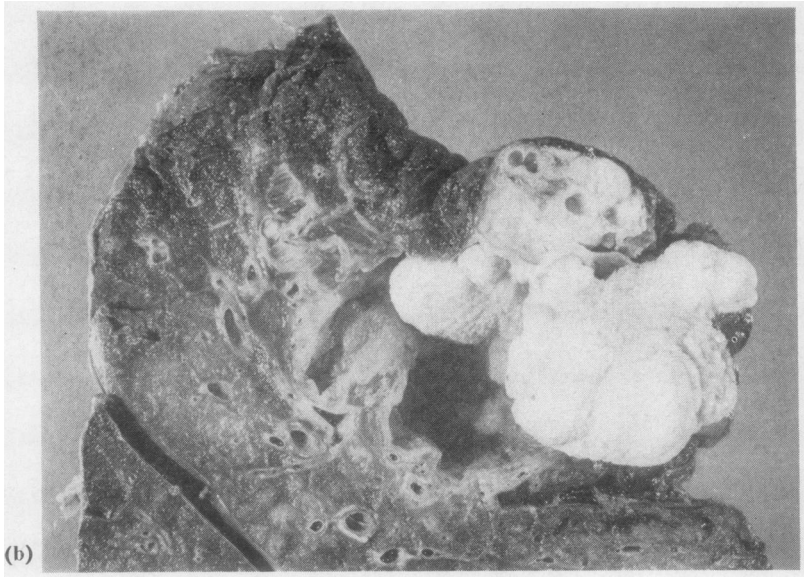
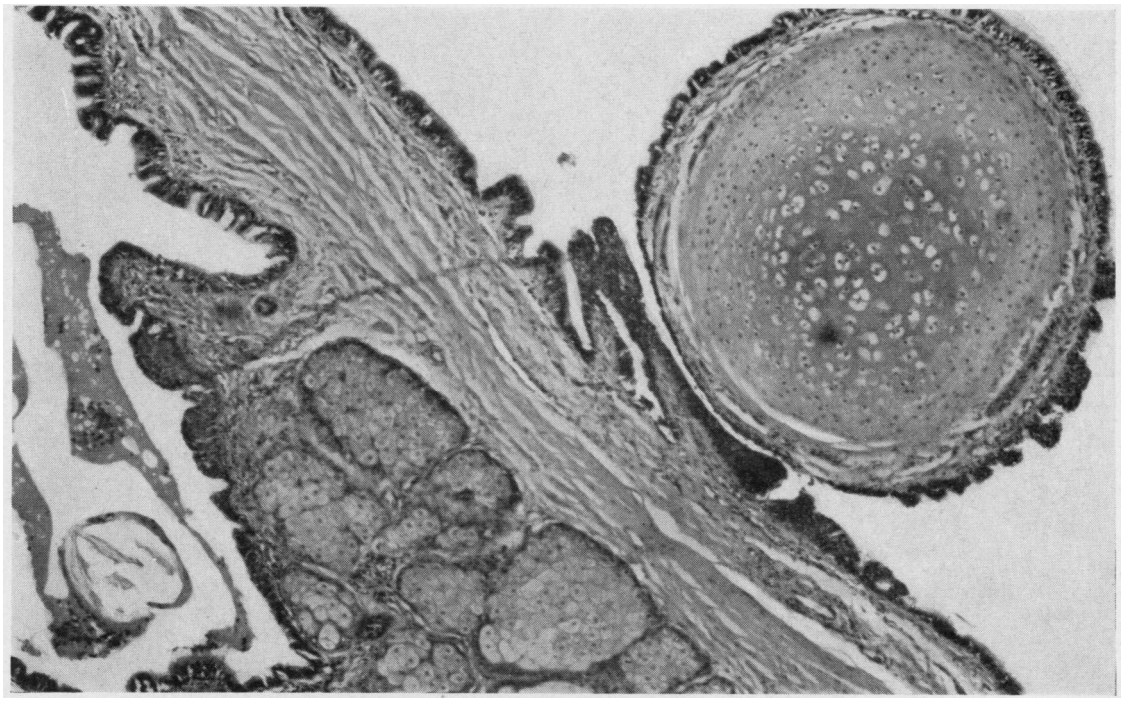
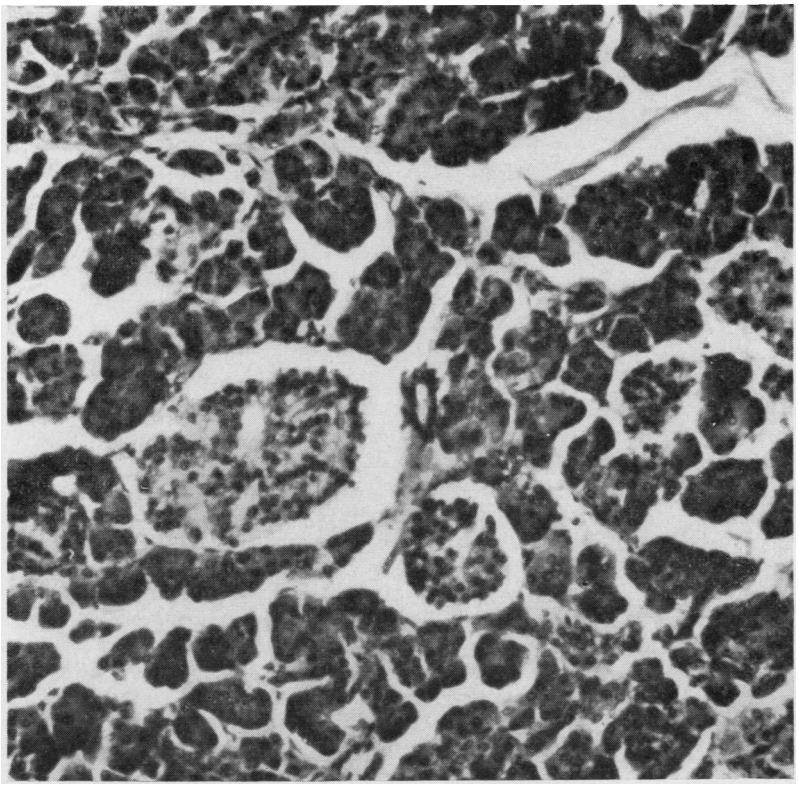


FIG. 5. (a) *Cross-section of the left lung. The intrabronchial tumour can be seen at the margin of the upper lobe and is clearly separated by a thin, air-containing space from the surrounding lung.* (b) *Enlargement of the upper part of Fig. 5a with the teratoma hanging on its pedicle from the dilated bronchus.* (c) *Enlargement of the lower part of Fig. 5a showing dilated bronchi and the yellow-white stellate areas (nodules) centred about dilated bronchioles.*





(a)



(b)

FIG. 6. (a) *Skin and appendages and primitive cartilage covered by respiratory type epithelium within the teratoma.* (*H. and E.* ×70.) (b) *Pancreatic tissue with islets of Langerhans within the teratoma.* (*H. and E.* ×570.)

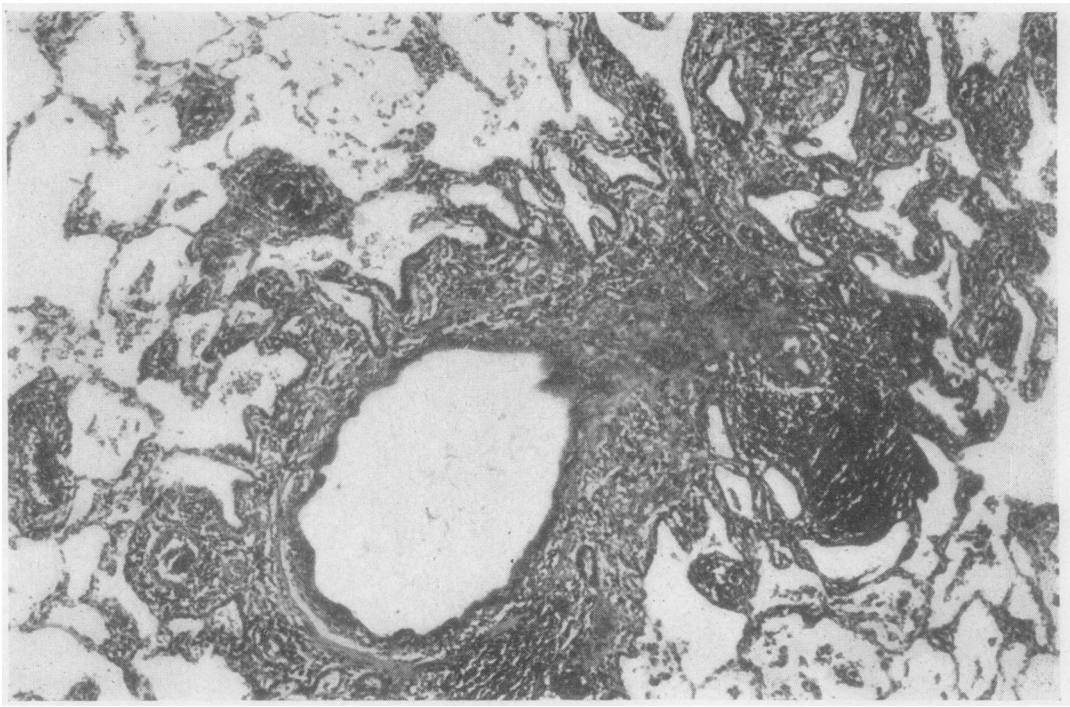


FIG. 6. (c) *Lung remote from the teratoma showing a terminal bronchiole with surrounding inflammatory reaction, mainly of lymphocytes and plasma cells. The alveolar walls are thickened and are lined by cubical epithelium.* (*H. and E.* ×235.)

raised the possibility that the lesion might be a mycetoma, the appearance being compatible with a fungus mass within a cavity (Monod, Pesle, and Labeguerie, 1952; Lodin, 1957; and Goldberg, 1962). Mycetomata, however, are usually thin-walled (Levin, 1956; Goldberg, 1962), unlike the present lesion, which had a thick irregular wall. They tend to be apical in position but may be situated adjacent to the mediastinum (Macartney, 1964). Hydatid cysts which have undergone partial rupture may sometimes present with a thin translucent crescent due to air between the pericyst and endocyst (Barrett and Thomas, 1952; Latham, 1953; Borrie, 1962), but they usually have well-defined margins. A crescent-shaped translucent area within a mass may be produced by a blood clot in a cavity (Ellis, Nathan, and Jones, 1961), a slough in a lung abscess (Kegel and Fatemi, 1961), and, theoretically, by any lung lesion which may cavitate, including neoplasm.

There was no doubt that the teratoma in the present case was endobronchial because its external surface was covered by bronchial mucosa and the mass lay inside a small bronchus which was grossly distended and formed an irregularly shaped cavity to which the tumour was attached at one point by a pedicle. The translucent crescent-shaped area within the irregular shadow was due to air in the lumen of the distended bronchus between the mass and the bronchial wall. This is a most unusual way for an endobronchial tumour to present radiologically, since the majority, which occur in the main bronchi, whether benign or malignant, produce collapse of the lung distal to the obstructed bronchus. The histological appearance of the teratoma is quite typical, especially the presence of pancreatic tissue, which is seen more commonly in teratomata situated in the thorax (Schlumberger, 1946).

A review of the previously published cases of intrapulmonary teratomata revealed that, although they were lying in the lung parenchyma, at least two of them showed some connexion with a bronchus. The tumour in the case reported by Trivedi *et al.* (1966) was attached by a pedicle to the lung, and through this the capsule of the tumour was connected with the middle lobe bronchus. Collier, Dowling, Plott, and Schneider

(1959) also described a case with a bronchial connexion. Their teratoma was cystic and lined by stratified squamous epithelium. This was continuous with the lining of the posterior segmental bronchus, which was also stratified squamous in character and merged proximally with normal respiratory epithelium. Radiologically, their case showed a cavity in the shadow of the mass which was inside the cystic teratoma, unlike the present case, where the crescent-shaped cavity was outside and around the tumour.

However, one case of teratoma, reported by Laffitte in 1937, was very similar to our case. This was a young woman aged 21 years who presented with chest pain, cough, and haemoptysis, and was found on radiological examination to have a round shadow in the left upper lobe. A needle was inserted into the left lung and lipiodol was injected into the superficial part of the mass. The contrast medium encircled the mass, forming a thin layer between it and the lung and then entered a small bronchus of the upper lobe. The mass was enucleated and a pencil-sized bronchus was found to open into the cavity which contained the mass. Histologically the mass showed the features of a teratoma and was covered by skin with hair follicles. It is obvious that the cavity left by the tumour was the lumen of a bronchus distended by the mass, but, unlike our case, the tumour itself was not invested by bronchial epithelium.

Theories have already been advanced to explain the origin of the mediastinal and intrapulmonary teratomata from the third pharyngeal pouch, which is the anlage of the thymus (Schlumberger, 1946). The migration of aberrant tissue from the third pharyngeal pouch along the developing bronchi gives rise to the intrapulmonary type of teratoma. Growth of the aberrant tissue forms a mass surrounded by lung which retains a bronchial connexion at one point. In the present case it is thought that the aberrant tissue giving rise to the teratoma expanded into the bronchus, instead of the lung parenchyma, which resulted in its endobronchial position. A similar hypothesis has been put forward to explain the difference between pure chondroma and chondromatous hamartoma (mixed tumour) of the lung (Bateson, 1967).

BRONCHIECTATIC CHANGES In addition to the bronchiectasis demonstrated bronchographically, which was 'follicular' on pathological examination, there was widespread bronchiolectasis. This bronchiolectasis produced the yellow-white stellate areas seen on macroscopic examination of the cut surface of the resected lung and showed up as the nodular opacities in the tomogram. This appears to be an unusual cause of nodular shadows in the lung and is not mentioned in the causes listed by Buechner (1959), Scadding (1952), and Schinz, Baensch, Friedl, and Uehlinger (1953).

The possibility of a connexion between the teratoma, follicular bronchiectasis, and widespread bronchiolectasis should be considered. It is very tempting to explain the ectasia of the bronchi and bronchioles from pressure on and occlusion of the left main bronchus (by the teratoma and/or enlarged hilar glands) at some stage during the development of the teratoma. However, Whitwell (1952) distinguished between follicular and atelectatic bronchiectasis, and this seems to exclude the explanation of the bronchiectasis as a result of extrinsic bronchial pressure, although marked enlargement of the hilar glands was a feature of the cases of follicular bronchiectasis in Whitwell's series. In our case the histological appearances of the bronchiectasis differed from the description given by Whitwell (1966) in the absence of marked fibrosis. The bronchiolectatic changes do not seem to have been reported previously either as a separate lesion or associated with either bronchiectasis or teratoma of the lung. It is possible that drainage of infected material produced by the teratoma may have been the cause of these changes.

We wish to thank Mr. C. Forrest, of the Pathology Photographic Department of the University of the West Indies, for the preparation of the illustrations and Mrs. Ivy Deans, *X*-ray Department, University Hospital, for typing the manuscript.

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